Case Report

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Toothache Caused by Sialolithiasis of the Submandibular Gland

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Sialolithiasis is the most frequent disease of the salivary glands, causing swelling and/or pain of the affected site. We report a 44-year-old woman who presented with severe pain in the lower left second molar region without swelling. Sialoliths on her left submandibular gland were confirmed by radiographic examinations. After robot-assisted sialoadenectomy, the pain did not recur but remained facial paralysis and unaesthetic scar.

Key Words: Hypesthesia; Salivary gland calculi; Salivary gland diseases; Submandibular gland; Toothache

INTRODUCTION

Non-odontogenic toothache refers to pain experienced without any clinically evident cause related to the teeth or periodontal tissues. Its common sources include myofascial, sinus/nasal mucosa, neurovascular, neuropathic, cardiac, somatoform, and systemic.[1] It is known that salivary gland diseases can cause non-odontogenic toothache,[2] but there are few reports about its symptoms or the characteristics of pain. Salivary stones or sialoliths are calcified structures or concretions located in the salivary glands. Sialolithiasis is a common salivary gland disorder characterized by the obstruction of the salivary secretion, accounting for approximately one-third of salivary gland disorders.[3] Typically, sialolithiasis presents a painful swelling of the gland at meal times when the gland is active.[4] This paper reports an uncommon case of sialolithiasis in the submandibular gland presenting intermittent spontaneous pain on mandibular molar area without swelling or pain on the salivary gland region, and discusses the related neurophysiological theory, the risks and benefits of the treatment of sialolithiasis.

CASE REPORT

A 44-year-old woman presented to the Department of Orofacial Pain and Oral Medicine of Yonsei University Dental Hospital (Seoul, Korea) complaining of stabbing pain in the mandibular left second molar, which occurs episodically 2-3 times a year from 3 years ago. She had heard of a radiopaque lesion on her left mandibular area on her panoramic radiography by a dentist of a local clinic. She visited other local clinics for the pain and heard that there was no specific abnormality with the tooth. The characteristics of her pain were intermittent and spontaneous with severe

intensity (numeric rating scale 9-10) which lasted about 10 minutes once it occurred. She explained that there were no other symptoms, such as swelling or fever. She had no other past medical or dental history other than having mandibular angle reduced by plastic surgery in 1994.

On dental examination there were no abnormal findings on her mandibular left second molar. But, on her panoramic radiography, a radiopaque material was observed on her left submandibular area as she heard before (Fig. 1). She was clinically diagnosed as sialolithiasis on left submandibular gland, which was presumed to be the origin of the pain. On mandibular computed tomography (CT), there were multiple radiopaque lesions on her submandibular gland and the affected gland appeared atrophied (Fig. 2). On her salivary gland scintigraphy, the uptake to the left submandibular gland was severely decreased and after the gustatory stimulation, no definite radiotracer extraction was noted (Fig. 3). Two sialoliths about 5 mm diameters (Fig. 4) were found during robot-assisted submandibular glandectomy after a referral to the Department of Oral and Maxillofacial Surgery. After the surgery, the pain of mandibular left second molar did not recur for 8 months, which was the latest follow-up, but postoperative complications including numbness on left facial area and unaesthetic scar of the surgical site occurred.

DISCUSSION

Most cases of sialolithiasis present with symptoms, such as pain and swelling in the submandibular or parotid region during mealtime. More than 80% of patients with sialoliths experience salivary gland swelling and 35% to 47% patients with sialoliths in submandibular gland suffer from pain.[5] But 3.1% of the patients have only pain without swelling as an isolated symptom.[6]

Somatic input from the face and oral structures is carried by way of the trigeminal nerve, entering directly in the brainstem in the region of the pons to synapse in the trigeminal spinal nucleus. It has been known that several primary sensory neurons must synapse with a single second-order neuron, which is known as convergence. Referred pain is a spontaneous heterotopic pain that is felt in an area innervated by a different nerve from the one that mediates the primary nociceptive input. The primary somatosensory innervation of the submandibular gland comes from the lingual nerve and those of mandibular molar teeth and gingiva are from inferior alveolar nerve, both of which are the subdivisions of mandibular nerve. It is conceivable that when nociceptive impulse of the submandibular gland excites the second-order neurons, the convergence in the trigeminal sensory nucleus may lead to confusion over the patient's pain site.[1] Therefore, clinician must consider a possibility of non-odontogenic toothache caused by salivary gland disease if a patient suffers from atypical tooth pain on lower molar area.

There are several methods to diagnose common salivary gland diseases: plain radiography, sialography, ultrasound, magnetic resonance imaging, CT, sialendoscopy, salivary gland scintigraphy, and fluorine-18 fluorodeoxyglucose

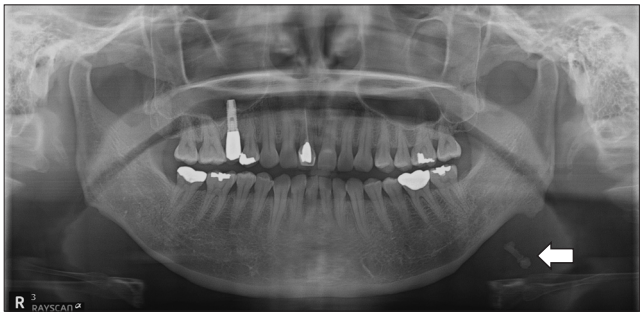

Fig. 1. Panoramic radiograph showed radiopaque lesions on left submandibular area (arrow).

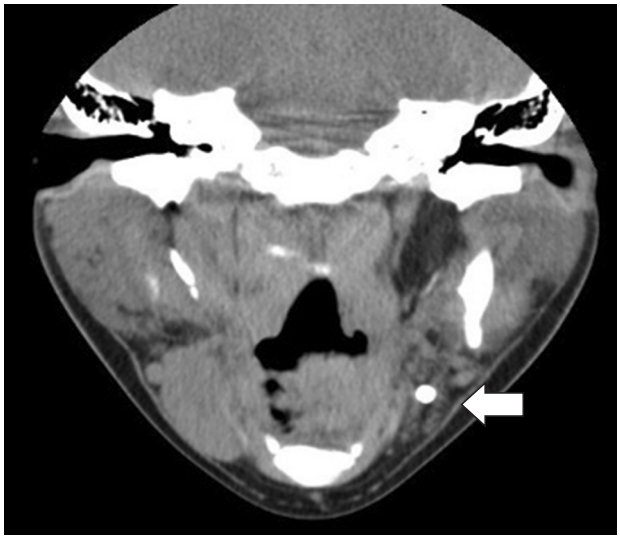

Fig. 2. Computed tomographic image showed atrophied submandibular gland on left side (arrow).

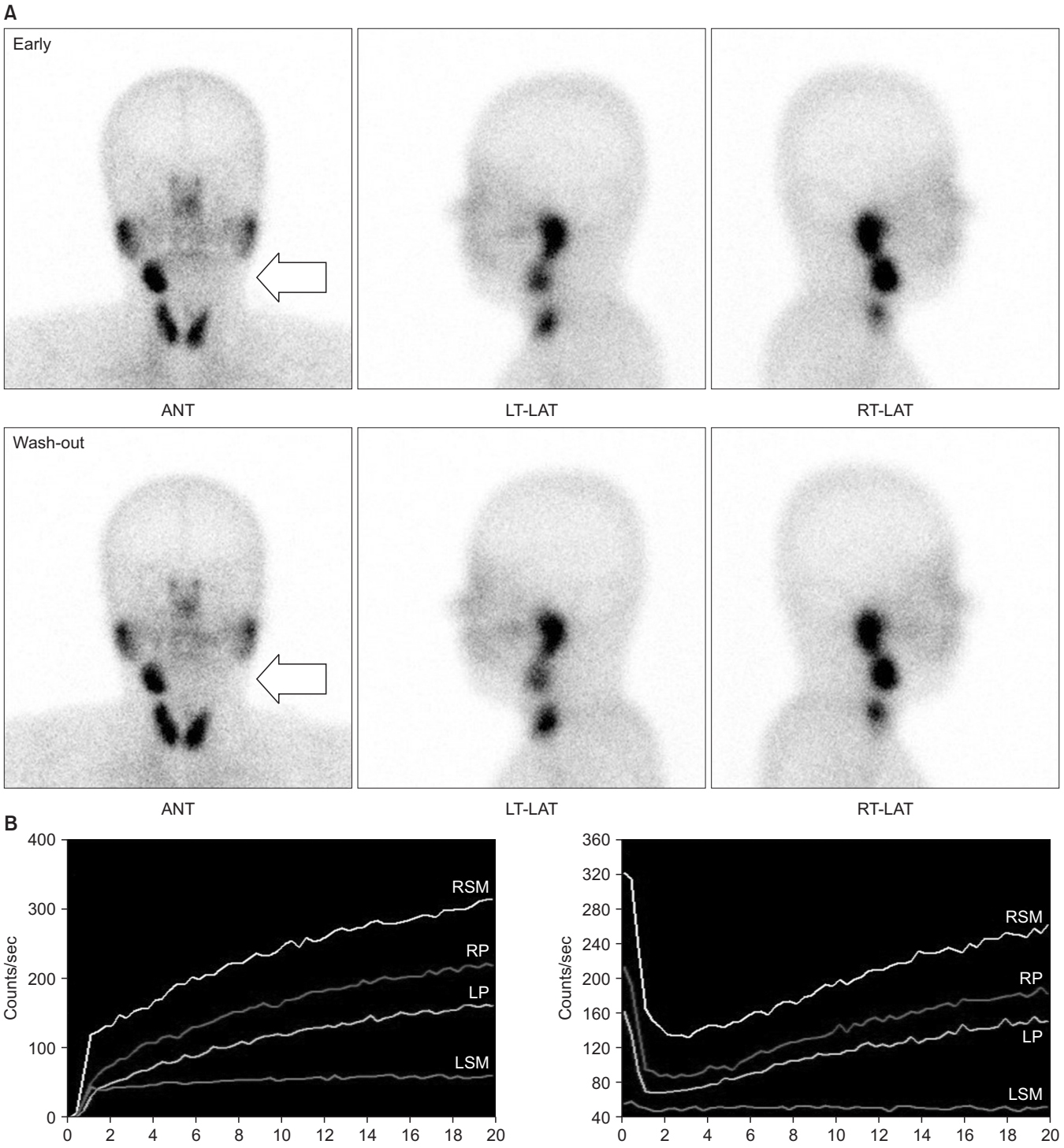

Fig. 3. (A) On salivary gland scintigraphy, severely decreased uptake to the left submandibular gland was observed (arrows). (B) Time-activity curves show a significant decrease of radioactivity of the left submandibular gland (LSM) before and after stimulation, comparing to other major salivary glands. ANT, anterior; LT_LAT, left lateral; RT_LAT, right lateral; RSM, right submandibular gland; RP, right parotid gland; LP, left parotid gland.

positron emission tomography. Plain radiography, such as panoramic view or mandibular occlusal view is an inexpensive and simple way of studying sialolithiasis in the salivary glands.[7] However, for small-sized or poorly calcified sialolithiasis or if radiopaque calcifications are superimposed on the mandible, further imaging modalities are needed.

Fig. 4. The affected submandibular gland was removed and two sialoliths were found inside the gland.

The treatment of choice for sialoliths associated with the submandibular gland is related to its location and size. A small or palpable sialolith in the anterior portion or the duct can be extracted intraorally by opening the orifice. Larger stones embedded in the hilum or the body of the submandibular gland may require more invasive approach, such as lithiotripsy, endoscopic techniques or sialoadenectomy.[8] Recently, minimally invasive surgical techniques are preferred to sialoadenectomy for some reasons. First, it is demonstrated that secretory function of the affected gland can recover after removal of the obstruction.[9] Su et al.[10] reported 17 patients showing glandular function recovery after removal of sialoliths by assessment of pre- and postoperative sialometric analysis and salivary gland scintigraphy. Second, sialoadenectomy can eradicate the obstructive salivary gland symptoms; however, at the same time, it may cause possible postoperative complications. Berini-Aytes and Gay-Escoda[11] studied postoperative complications in 206 submandibular gland excisions. Neurological complications (33 cases, 16.0%) and unaesthetic scars (10 cases, 4.8%) were observed. Facial nerve (11.6%) was the most commonly affected nerve followed by the lingual nerve (4.4%) and hypoglossal nerve (3.4%). Immediate postoperative problems of sialoadenectomy, such as hemorrhage and infection are usually transient rather than life-threatening, and may only delay hospital discharge. In contrast, iatrogenic nerve injuries cause the greatest concern both to the surgeon and the patient, because they can be permanent and diminish the patient's quality of life. McGurk et al.[12] reviewed sixteen papers reporting submandibular gland adenectomy with associated complications. Overall, temporary injury to the facial nerve was approximately 9.6%, with permanent injuries at 3.3%. Injury to the lingual and hypoglossal nerves was less common (less than 4.0%) but half of these may be permanent. Also, a scar may remain one of the major problems after sialoadenectomy which is unavoidable and can cause significant implications for the patient. It can cause discomfort, tightness or even pain and psychological problems.[13] Therefore, early diagnosis & removal of sialolith with minimally invasive technique may be helpful to prevent the salivary glands from loss of function and postsurgical sequelae.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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